DISCUSSION

Hypnic Headache is a benign nocturnal headache which predominantly affects elderly people. This case illustrates some classic features of the syndrome. Evers and Goadsby[1] reviewed the 71 reported cases giving us the clearest picture of the syndrome to date. 37% were male and 63% female. Mean age of onset was 63 +/- 11 years (range 36-83). Headache was described as: Moderate - 67%, Severe - 31%; Dull - 57%, Throbbing/Pulsating - 38% and Sharp/Stabbing - 5%; Diffuse - 57%, Frontotemporal - 42%, Posterior - 1.6%. Average duration was 67 +/- 44 minutes (range 15-180). Onset was 60 - 120 minutes after falling asleep in 77%. Nausea was reported in 19%. The pathophysiology of HHS is currently theoretical, but associations with the sleep/wake cycle and circadian rhythms form the basis for theories of its nature. Polysomnography has revealed the onset of hypnic headaches may be associated with REM sleep.[2] It may be that inactivation of antinociceptive structures, e.g. dorsal raphe, during REM mediates the headache.[3]

Commonly patients experience the headache at a predictable time each night, suggesting a link with the circadian rhythm, which is orchestrated by the suprachiasmatic nuclei in the hypothalamus (also involved in antinociception). These nuclei produce, among others, melatonin, an important mediator of circadian rhythm. With advancing age the function of the hypothalamus, and thus melatonin secretion, is impaired.[4] This could also be involved in the pathogenesis of hypnic headache. Lithium is believed to increase melatonin levels[5] and may explain its mode of action. However, undoubtedly it is more complex than any one of these associations as many different drugs have been tried with variable success. Lithium remains the most effective but is often limited by side effects and interactions, and requires monitoring of plasma concentrations to avoid toxicity. Other reported treatments include indomethacin, caffeine, verapamil, prednisolone, gabapentin, melatonin, and acetazolamide.

Awareness of benign headaches is important to avoid unnecessary investigation but it must be stated that brain imaging and routine biochemical/haematological investigations are usually indicated when presented with new onset headaches in the elderly.

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The authors have no conflict of interest

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Myopathy, hypokalaemia and pica (geophagia) in pregnancy

Editor,

Pica describes the persistant ingestion of nonnutritive substances.[1] Geophagia describes pica of clay.[2] We present a case of geophagia resulting in hypokalaemic myopathy.

Case History: A 29 year Gravida 3 Para 2 presented to a tertiary referral centre in Cape Town, South Africa, at 30^{+4} weeks gestation. She gave a two week history of photophobia, vomiting and weakness of the left side of her body. No other symptoms were reported. Fetal movement was reported to be normal. Her two previous pregnancies, in 1997 and 2001, were uneventful and resulted in normal vaginal deliveries at term. She had no significant medical or family history, was not on medication and did not report any allergies. She was a non-smoker and non-drinker. She was from the coloured community in Cape Town. She was a single mother, lived in an informal dwelling settlement, and had no monthly income.

She booked at 22 weeks gestation and an anomaly scan reported no fetal abnormality. Her booking Body Mass Index was 33, BP 110/85 mmHg, Hb 9.3g/dl, blood group A+ve, no abnormal antibodies, VDRL negative and HIV negative. Her pregnancy was uneventful up until presentation at hospital.

Examination of the cardio-vascular, respiratory and gastro-intestinal systems was normal. Neurological assessment of the central nervous system was normal. Proximal muscle strength was reduced bilaterally with muscle groups demonstrating 4/5 strength. Biceps and patellar reflexes were reduced and plantar reflexes were normal. Sensation was normal. The provisional diagnosis was a myopathic process of unknown aetiology.

Haematological investigations showed a Hb of 9.9g/dl, WCC 11.2×109/L, Platelets 391×109/L and an ESR of 73mm in 1 hour. Biochemistry showed a sodium of 145mmol/L, potassium 1.5mmol/L, urea 2.2mmol/l, and creatinine 116μmol/L. Liver function tests were also abnormal. Her creatine kinase was 9920U/L. Further investigations as an in-patient included and EMG, MRI scan of brain and muscle biopsy. The EMG reported features in keeping with a myopathy. The MRI scan and muscle biopsy were normal. Biophysical assessment of the fetus was reassuring.

Further questioning of the mother revealed that throughout the pregnancy she had regularly been eating clay from outside

her house. It was impossible to accurately quantify the amount eaten. The patient's symptoms slowly responded to intravenous and oral potassium supplementation over 14 days. Her liver function and renal function returned to normal. She was discharged 18 days after admission to hospital. Her care, for the remainder of her pregnancy, was in the community. Further follow-up information is not available. The patient did not have a telephone land line nor a cellphone. A request was made to the community services to follow-up the patient. On receiving the address, we were told that they only went into the patient's district with a police escort and our request was not justified.

DISCUSSION

The aetiology of pica is not known. Theories on pica range from nutritional deficiencies and psychological problems to obsessive-compulsive behaviour and specific brain lesions.[3,4] Pica can cause a number of serious conditions including iron-deficiency anaemia, bowel obstructions and perforations, lead poisoning, and helminthnic infestations.[5] This is only the second report in the literature of geophagia causing hypokalaemic myopathy in pregnancy.[6] The pathophysiology, it is suggested, is that clay binds to potassium in the gut. This leads to increased intestinal excretion of potassium, resulting in hypokalaemia.[7] It appears that the effect is dose dependent on the amount of clay ingested.

Unfortunately, our patient was lost to follow-up. This also occured in the other reported case.[6] The long-term maternal, fetal and neonatal outcomes in the severely hypokalaemic mother, would be of interest.

The author has no conflict of interest.

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Atypical presentation of HIV in a pregnant patient

Editor,

We report a case of Kaposi's sarcoma (KS) of the epiglottis in a pregnant lady who presented with stridor. It is very rare for stridor caused by laryngeal KS to be the initial presentation of HIV infection in a pregnant patient that has not been previously reported.

Case Report: A 33-year-old, 13 weeks pregnant lady presented with shortness of breath and noisy breathing. She also had odynophagia, bilateral neck swelling, sore throat and night sweats for the past 3 days. She had a discharging ear and a chronic non-productive cough for 3-6 weeks. Distaclor, commenced initially, was discontinued when she was confirmed to be pregnant. She was on nystatin mouthwashes for her oral thrush. She was a non-smoker and took alcohol occasionally. There was no history of intravenous drug abuse or other risk factors for HIV infection.

On examination she was pyrexic, had inspiratory stridor, tachycardia and tachypnoea. Oral cavity and oropharynx examination revealed extensive candidiasis.

Flexible nasoendoscopy revealed a very large, oedematous and inflamed epiglottis with extensive white patches. The epiglottic swelling was so large that the vocal cords could not be visualized and only the posterior portion of the arytenoids was seen. Neck examination revealed bilateral cervical lymphadenopathy. Lateral soft tissue X-ray of the neck revealed an enlarged epiglottis and a normal trachea. Chest X-ray was clear. Full blood count showed WCC - 6.1×10^9/L, Hb - 13.2 g/dl and platelets – 215×10^9/L. Routine blood tests and viral serology was normal. Her CD4 count was 40/mm^3.

A provisional diagnosis of severe fungal / bacterial epiglottitis was made. The treatment regime included high dose of intravenous fluconazole, cefuroxime, metronidazole and nystatin mouthwashes. An HIV test was positive which was again confirmed on retesting.

An ultrasound scan revealed an anembryonic and nonviable pregnancy. After discussion with the patient she had a medical evacuation of the pregnancy with mifepristone.

Anti-retroviral therapy and prophylaxis with co-trimoxazole, azithromycin and dapsone was commenced. Two weeks later on review with flexible nasoendoscopy, the epiglottis still appeared inflamed and grossly swollen. A CT scan of the neck and upper thorax showed a 4 cm swelling of the epiglottis extending down into the aryepigolttic folds and into the vestibule. Bilateral cervical lymphadenopathy was also noted on the CT (*Fig 1*).

In view of her slow recovery, a microlaryngoscopy and biopsies of the epiglottis were performed to reach a firm diagnosis. The histopathology revealed necrotic inflammatory tissue with ulceration and dense proliferation of anastomosing vascular channels. A tentative diagnosis of KS was made. The specimen was sent to a tertiary referral centre for human herpes virus 8 (HHV8) staining which proved positive. The diagnosis was finally confirmed when the samples were sent to the national center of KS pathology.

